European Review for Medical and Pharmacological Sciences 2012; 16: 1891-1894

A rare location of benign osteoblastoma: case study and a review of the literature

R. CALTABIANO[3], A. SERRA[1], M. BONFIGLIO[1], N. PLATANIA[2], V. ALBANESE[2], S. LANZAFAME[3], S. COCUZZA[1]

[1]Otolaryngology Section, Department of Medicine and Surgery, University of Catania, Italy
[2]Department of Neurosurgery, Azienda Ospedaliero-Universitaria G. Rodolico, Catania, Italy
[3]Department G.F. Ingrassia, Section of Anatomic Pathology, University of Catania, Italy

Abstract. – Osteoblastoma is a solitary, benign bone tumor that is rarely localized in the frontal sinus. It consists of hypocellular mineralized tissue that may form large masses or irregular trabeculae. A 31 year old man came to our attention with a 7 month history of diplopia, photophobia, frontal headhaches and progressive exophthalmos with proptosis of the left eye. The patient was submitted to computed tomography (CT) which allowed to appraise the extension of the lesion. The mass expanded inside the left frontal sinus and the upper ethmoidal cells invading the left orbital roof. Considering the extension of the tumor, the site and the connections with contiguous structures, a combination of endoscopic endonasal technique with intraorbital approach was performed. At histological examination typical features of benign osteoblastoma were observed. The sites of predilection for the tumor are the long bones, vertebral column, and small bones of hands and feet. Its occurrence in the skull and jaw bones is relatively rare and represents only 15% of all osteoblastomas. To our knowledge, only 5 cases of osteoblastoma of the frontal sinus have been previously reported in the English-language literature. This report describes a case of benign osteoblastoma in a rare site, namely, the frontal sinus with particular attention about the differential diagnosis and the treatment.

Key Words:
Osteoblastoma, frontal sinus, case report, review, histopathology.

Introduction

Osteoblastoma is a solitary, benign bone tumor that is rarely localized in the frontal sinus. It consists of hypocellular mineralized tissue that may form large masses or irregular trabeculae. This mineralized matrix contains numerous hyperchromatic osteoblasts and occasional osteoclast-like giant cells in a richly vascular fibrous stroma. It is most common in the vertebrae of children and young adults. Only 5 cases of osteoblastoma of the frontal sinus have been previously reported in the English literature[1-5]. This report describes a case of benign osteoblastoma in a rare site, namely, the frontal sinus, with particular attention about the differential diagnosis and the treatment.

Case Report

A 31 year old man came to our attention with a 7 month history of diplopia, photophobia, frontal headhaches and progressive exophthalmos with proptosis of the left eye. He reported that 8 months before he had undergone a right frontal craniotomy and cranioplasty to remove a right frontal sinus lesion. Postoperative computed tomography (CT) carried out 2 months after surgery showed a hypodense mass in the left frontal sinus. During the following 5 months the patient experienced a gradual worsening of symptoms and underwent magnetic resonance imaging (MRI) which demonstrated a bulky lesion, with isointense signal on T1-weighted scans and with a heterogeneous enhancement after the contrast medium injection, extending into the frontal sinus, the ethmoidal labyrinth and the nasal cavity of the left side (Figure 1). Ophtalmological examination showed exophtalmos, proptosis and mild abduction deficit in the left eye. Diplopia was present in primary position and in all gaze directions. Fundus examination of the left eye showed convoluted vessels. Normal visual acuity, no abnormalities of anterior segment and normal light reflex in both eyes were reported. General physical examination didn't show any abnormalities and blood exams depicted a mild increase of white blood cell count and lactate dehydrogenase. The patient was submitted to CT which allowed to appraise

Corresponding Author: Salvatore Cocuzza, MD; e-mail: scocuzza@unict.it - salvatorecoc@yahoo.it

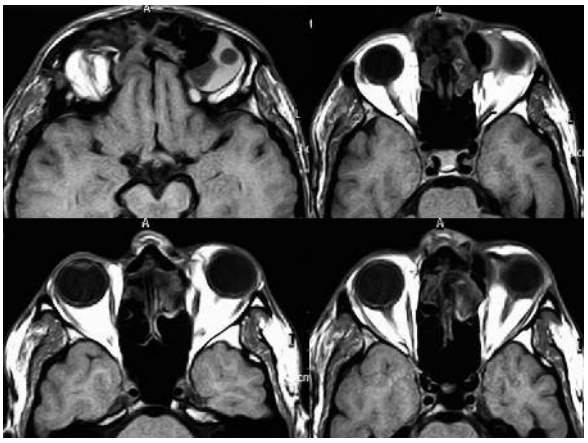

Figure 1. T1 weighted axial MRI scans showing a huge orbito-ethmoidal disomogeneous lesion on the left side.

the extension of the lesion. The mass expanded inside the left frontal sinus and the upper ethmoidal cells invading the left orbital roof (Figure 2). Moreover, CT showed the narrow contact between the lesion and medial rectus muscle in proximity to its insertion on the globe. Considering the extension of the tumor, the site and the connections with contiguous structures, a combination of endoscopic endonasal technique with intraorbital approach was performed. The orbital structures were preserved and a complete macroscopic removal of the mass was accomplished. At gross examination, the lesion showed a red granular appearance. Microscopically, the tumor presented anastomosing bony trabeculae rimmed with a single layer of osteoblasts (Figure 3). The intertrabecular spaces were arranged loosely and contained occasional spindle cells and capillaries. Considering all these features, a diagnosis of osteoblastoma was performed. Postoperative CT assessment confirmed the complete removal of the lesion (Figure 4). The patient recovered well and local symptoms resolved in two months. The patient was free of recurrence at 24 month follow-up visit.

Discussion

The osteoblastoma accounts for approximately 1% of all primary bone tumors[6] Jaffe and Mayer[7] first described the osteoblastoma in 1932 when they reported a case of an "osteoblastic osteoid tissue–forming tumor" that arose in the fourth metacarpal bone of a 15-year-old female. The sites of predilection for the tumor are the long bones, vertebral column, and small bones of hands and feet. Its occurrence in the skull and jaw bones is relatively rare and represents only 15% of all osteoblastomas[8-9]. The tumor shows a sex predilection for males and constitutes less than 1% of all tumors of the maxillofacial region[10]. Primary osteoblastoma of the frontal sinus, however, is rare. We are aware of only 5 published cases, all single case reports[1-5]. The radiographic appearance is not specific and extremely variable. Depending on the degree of calcification in the lesion, it may appear radiolucent or semiradiolucent with radiopaque mottling and a well-demarcated margin. As in our case, CT is

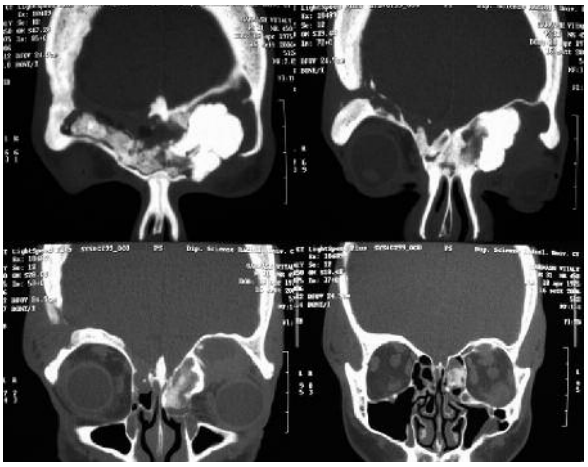

Figure 2. Coronal bone CT scans showing the extension of bony lesion inside the left frontal sinus and upper ethmoidal cells and the wide destruction of the left orbital roof.

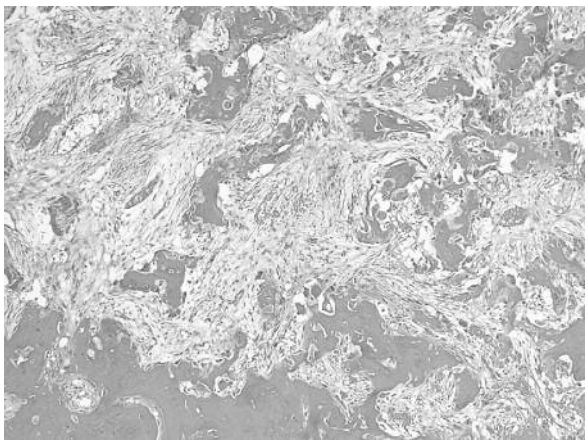

Figure 3. Markedly well vascularised fibrous connective tissue containing osteoid and bone trabeculae surfaced by numerous osteoblasts (hematoxylin-eosin stain, original magnification 50×).

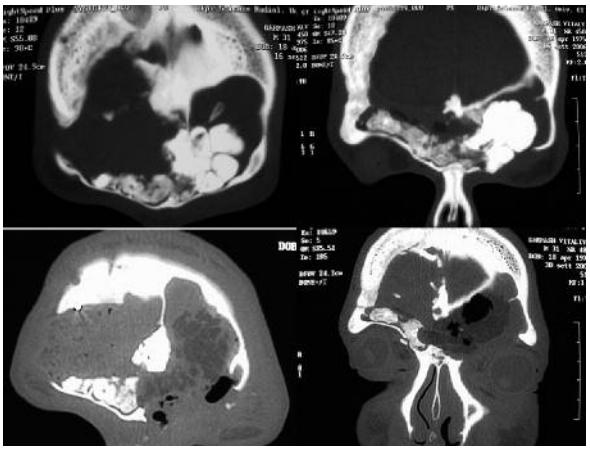

Figure 4. Upper row: preoperative CT coronal bone scans showing the extension of the lesion. Lower row: postoperative CT coronal bone scans showing the complete removal of the left orbito-ethmoidal lesion after combined endoscopic trans-nasal ethmoidal and supraorbital neuronavigation-assisted approach.

a useful adjunct to the diagnosis and is of great value in determining the extent of the tumor as well as the degree of calcification. Rare cases have a nidus associated with sclerosis as seen in osteoid osteomas. According to the literature, an osteoid osteoma should demonstrate a radiographic nidus that is less than 1 cm in size, whereas an osteoblastoma should measure more than 2 cm in greatest dimension[11-12]. Histologically, the most pathognomonic features are the presence of islands of osteoid tissue undergoing varying degrees of calcification and considerable osteoblastic activity in a loose, well-vascularized stroma. Osteoblastoma, therefore, is a benign, slow growing neoplasm of bone characterized by the proliferation of numerous, plump osteoblasts forming osteoid and bone trabeculae set in a markedly well-vascularized fibrous connective tissue stroma. The stroma varied from loosely arranged collagen fibers interspersed with spindle-shaped fibroblasts to more densely arranged collagen fibers. The stroma contained numerous endothelium lined, blood filled vascular channels along with extravasated erythrocytes. The trabeculae are rimmed by plump osteoblasts exhibiting oval to round-shaped basophilic nuclei and an abundant eosinophilic cytoplasm. Areas of bone remodelling with adjacent osteoclasts are also evident. In some cases, large sheets of calcified osteoid are noted. The osteoblasts do not display atypical cytologic features or mitotic figures. The presence of atypia is cause for careful differential diagnosis of this lesion from osteosarcoma and osteoblastic lesions of uncertain malignant potential. Differential diagnosis include the osteoid osteoma, aggressive osteoblastoma and osteosarcoma. The similarity of osteoblastomas to osteoid osteomas was already reported in 1954 by Dahlin and Johnson[13]. The osteoma is the most common tumor of the paranasal sinuses[14]. The most common locations are frontal, ethmoid, maxillary, and sphenoid sinuses in that order. Osteomas may have central areas of radiolucency that in some cases correlate with woven bone with microscopic features indistinguishable from osteoblastoma[15]. Unlike osteoid osteoma, osteoblastoma has a predilection for involving the spine. The tumor in this case was differentiated from osteoid osteoma, which is histologically characterized by a central nidus of osteoid tissue and a constant perifocal osseous reaction, a scanty stromal reaction, rare multinucleated giant cells, and less vascularisation than the benign osteoblastoma. The biologic behavior of benign osteoblastoma is not constant, and it shows wide variations. Although considered benign, some lesions show aggressive behaviour and rare malignant transformation into histologically typical osteosarcoma are reported[16]. It is critical to differentiate between the aggressive osteoblastoma and the conventional osteoblastoma, as the former tumor shows a far greater likelihood of recurrences In some cases, aggressive osteoblastomas have led to patient death, exhibiting a pattern of tumor growth similar to a low-grade osteosarcoma. The aggressive osteoblastoma and the conventional osteoblastoma both frequently occur in the vertebral column, tibia, femur, and skull. The aggressive osteoblastoma may be larger at the time of diagnosis. The histologic characteristics of the aggressive osteoblastoma overlap somewhat with the conventional osteoblastoma, although the aggressive osteoblastoma contains plump epithelioid-appearing osteoblasts, lining the osteoid trabeculae. The osteoblasts are approximately twice the size of osteoblasts seen in a conventional osteoblastoma with an abundant eosinophilic cytoplasm and large nuclei with prominent nucleoli. In some cases, the aggressive osteoblastoma has more multinucleated giant cells of the osteoclast type and more abundant atypical osteoid[17]. Osteosarcoma is more likely to exhibit anaplasia with atypical mitoses, a high rate of mitotic activity, infiltrative margins with permeation of the surrounding trabecular bone, lace-like or streamer osteoid, production of cartilage, and sheets of malignant cells without osteoid production.

Treatment of osteoblastoma with local conservative excision and curettage should be adequate. The surgical approach for complete removal of the tumor has always been variable, depending on the tumor size and site, although in some cases the access had been potentially severely mutilating[18]. The osteoblastoma shows a significant tendency to recur (13.6%), which calls for careful surgical management and follow-up. Recurrence was attributed to inadequate or conservative initial treatment, including incomplete local curettage or partial resection of the tumor and not to inherent biologic behavior. However, even with a limited conservative approach, complete resolution or regression have occurred and, in some cases, spontaneous regression took place after biopsy. When a conservative approach is used, close long-term follow-up of the case is paramount. In the current case, no sign of recurrence was observed 4 months postoperatively.

References

1) Garstecka A, Mackiewicz-Nartowicz H, Szukalski J. Benign osteoblastoma of the frontal sinus. Otolaryngol Pol 2004; 58: 649-652.
2) Parodi MB, Iustulin D, Isola V. Partial ablation of benign osteoblastoma: a case report. Metab Pediatr Syst Ophthalmol 1993; 16: 43-45.
3) Szlezak L, Przybora L, Markowska M. Benign osteoblastoma of the ethmoid labyrinth, frontal sinus and orbit. Klin Oczna 1975; 45: 247-251.
4) Korytowski J, Dulczyński B, Przybora L. Case of frontal sinus osteoblastoma. Otolaryngol Pol 1974; 28: 575-579.
5) Vensi E. Multiple benign osteoblastoma of the femur and frontal sinus. Ann Laringol Otol Rinol Faringol 1965; 64: 768-770.
6) Dorfman HD, Czerniak B. Bone tumors. St Louis: Mosby: 1998; pp. 85-114.
7) Dahlin DC, Johnson EW Jr. Giant osteoid osteoma. J Bone Joint Surg Am 1954; 36: 559-572.
8) Huvos AG. Bone Tumors. Diagnosis, Treatment and Prognosis (ed 2). Philadelphia, PA, Saunders, 1991.
9) Lucas DR, Unni KK, McLeod RA, O'Connor MI, Sim FH. Osteoblastoma: clinicopathologic study of 306 cases. Hum Pathol 1994; 25: 117-134.
10) Haug RH, Hauer C, De Camillo AJ, Araneta M. Benign osteoblastoma of the mandible: Report of a case. J Oral Maxillofac Surg 1990; 48: 743-748.
11) Unni KK. Dahlin's bone tumors. General aspects and data on 11,087 cases. Philadelphia: Lippincott-Raven; 1996, p. 121-142.
12) Frassica FJ, Waltrip RL, Sponseller PD, Ma LD, McCarthy EF Jr. Clinicopathologic features and treatment of osteoid osteoma and osteoblastoma in children and adolescents. Orthop Clin North Am 1996; 27: 559-574.
13) Jaffe HL, Mayer L. An osteoblastic osteoid tissue–forming tumor of a metacarpal bone. Arch Surg 1932; 24: 550-564.
14) Eller R, Sillers M. Common fibro-osseous lesions of the paranasal sinuses. Otolaryngol Clin North Am 2006; 39: 585-600.
15) Dorfman HD, Czerniak B. Bone Tumors. St Louis, MO: The CV Mosby Co; 1998: ix, 1261.
16) Dorfman HD, Weiss SW. Border line osteoblastic tumors: Problems in the differential diagnosis of aggressive osteoblastoma and low-grade osteosarcoma. Semin Diagn Pathol 1984; 1: 215.
17) Mitchell ML, Ackerman LV. Metastatic and pseudomalignant osteoblastoma: a report of two unusual cases. Skeletal Radiol 1986; 15: 213-218.
18) Guest PG, Juniper RP. Osteoblastoma: a case report and description of the access used to the retromaxillary area. Br J Oral Maxillofac Surg 1991; 29: 333-335.